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Published in:
Journal of Intellectual Disability Research

DOI:
10.1111/jir.12679

Publication date:
2019

Document Version
Author accepted manuscript

Link to publication in ResearchOnline

Citation for published version (Harvard):
Pennington, M, Ring, H, Howlett, J, Smith, C, Redley, M, Murphy, C, Hook, R, Platt, A, Gilbert, N, Jones, E, Kelly, J, Pullen, A, Mander, A, Donaldson, C, Rowe, S, Wason, J & Irvine, F 2019, 'The impact of an epilepsy nurse competency framework on the costs of supporting adults with epilepsy and intellectual disability: findings from the EpAID study', *Journal of Intellectual Disability Research*, vol. 63, no. 12, pp. 1391-1400. https://doi.org/10.1111/jir.12679

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Download date: 05. May. 2023

Title

The impact of an epilepsy nurse competency framework on the costs of supporting adults with epilepsy and intellectual disability. Findings from the EpAID study.

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Running Head

Cost of the epilepsy nurse framework

Acknowledgements

This paper presents independent research commissioned by the National Institute for Health Research (NIHR) under its Health Technology Assessment Programme (Grant 10/104/16 PI Dr H Ring). During the time that this research was carried out, H Ring also received support from the NIHR Collaboration for Leadership in Applied Health Research and Care (CLAHRC) East of England at Cambridgeshire and Peterborough NHS Foundation Trust. The views expressed are those of the authors and not necessarily those of the NHS, the NIHR or the Department of Health. We are grateful to the participants and to their family and paid carers for the time they generously gave to this research.

Conflicts of interests

HR has received lecture fees from UCB and Eisai. MP has received consultancy fees from Merck. None of the other authors have any conflicts of interest to disclose.

We confirm that we have read the Journal's position on issues involved in the ethical publication and affirm that this report is consistent with those guidelines.

Abstract

Background: The development of a nurse-led approach to managing epilepsy in adults with an intellectual disability offers the potential of improved outcomes and lower costs of care. We undertook a cluster randomised trial to assess the impact on costs and outcomes of the provision of intellectual disability nurses working to a designated epilepsy nurse competency framework. Here, we report the impact of the intervention on costs.

Method: Across the UK, 8 sites randomly allocated to the intervention recruited 184 participants, 9 sites allocated to treatment as usual recruited 128 participants. Cost and outcome data were collected mainly by telephone interview at baseline and after six months. Total costs at six months were compared from the perspective of health & social services, and society, with adjustments for pre-specified participant and cluster characteristics at baseline including costs. Missing data was imputed using Multiple Imputation. Uncertainty was quantified by bootstrapping.

Results: The intervention was associated with lower per participant costs from a health & social services perspective of -£357 (2014/15 GBP) (95% CI -£986, £294) and from a societal perspective of -£631 (95% CI -£1,473, £181). Results were not sensitive to the exclusion of accommodation costs.

Conclusions: Our findings suggest that the competency framework is unlikely to increase the cost of caring for people with epilepsy and intellectual disability and may reduce costs.

Key words

Epilepsy, cost, intellectual disability, management, (name removed for blinded review), competency framework

Background

Epilepsy affects around a quarter of adults with intellectual disability (ID), a group numbering nearly one million in England (McGrother et al. 2006). A study of adults with ID in Scotland demonstrated a point prevalence of epilepsy in that sample of 34% (Kinnear et al. 2018). For comparison, the pooled estimate for the point prevalence of epilepsy in the general population, in high income countries, is 5.49 per 1000 (CI 4.16–7.26) (Fiest et al. 2017). Epilepsy treatment outcomes are worse and management costs are higher in this group, possibly reflecting both disease severity and the challenges of managing health conditions in people with ID (Kerr, 2011). The challenges of managing epilepsy in people with ID and the need for innovative and holistic approaches to coordinating care were recognised in recent reports from Public Health England (Marriott et al. 2014) and others (Kerr et al. 2014; MENCAP 2007).

Clinical guidelines in the UK recommend a key role for specialist epilepsy nurses in the management of epilepsy (Kerr et al. 2014; NICE 2012). However, nurses with specialist epilepsy experience are rare and consequently access is very limited (Reuber et al. 2008). There is a lack of a robust evidence base on the cost-effectiveness of epilepsy nurse specialists to support the expansion of provision. Evidence to date suggests that epilepsy nurse specialists have limited impact on clinical outcomes for patients but may reduce care costs (Johnson et al. 2010; Warren et al. 1998).

The UK Epilepsy Specialist Nurse association has published a set of guidelines to support the delivery of epilepsy care in people with ID - the Learning Disability Epilepsy Specialist Nurse Competency Framework (Doherty et al. 2013). The framework supports clinical practice and professional development relating to the management of epilepsy by nurses with a range of experience in the treatment of epilepsy in people with an ID, from novice to expert – it is not designed just to be used by experienced

epilepsy nurses. This is important as the majority of nurses already working in services that manage people with ID and epilepsy are not epilepsy specialist nurses.

The framework was evaluated in (name removed for blind review), a cluster randomised controlled trial across 17 centres in the UK. The trial protocol (citation removed for blind review) and full trial report (citation removed for blind review) have been previously reported. In this paper we report the impact of the competency framework on the cost of caring for people with epilepsy and ID. Effectiveness and cost-effectiveness of the framework will be reported in a later publication.

Methods

Study design and participant recruitment

(Name removed for blinded review) was implemented as a cluster randomised trial to reflect the likelihood that nurses within a community team were likely to share duties and to influence each other's practice, and the practical difficulty in delivering 'treatment as usual' after having received specialist training. Sites were randomised to treatment or control in blocks of two or more and after participant recruitment. Whilst nurses could not be blinded to treatment allocation, people with epilepsy and their carers were not explicitly informed. Outcome and cost data were collected by research assistants blinded to treatment allocation. Initial plans to recruit 32 participants at each of 12 sites were modified to 20 participants at each of 16 sites after recruitment challenges. All participants were recruited from secondary care adult community ID teams located in both inner city and more rural areas in England, Scotland and Wales.

The trial was powered on the primary outcome, which was the Epilepsy and Learning Difficulties Quality of Life (ELDQoL) (Buck et al. 2007) seizure severity subscale, requiring 16 participants per site to provide 90% power to detect a difference of 3.6 at a

one sided significance level of 0.025. Participants were eligible for recruitment if they were aged 18-65, IQ of 70 or less, a diagnosis of epilepsy with at least one seizure in the previous six months, and had care input from a nurse in the Community Intellectual Disability Team. Participants with a rapidly progressing physical or mental illness, and those with drug or alcohol dependence were excluded.

Consent was obtained in face to face interviews with the participant, or where the potential participant lacked capacity to consent, assent was obtained from the primary carer under the provisions of the Mental Capacity Act (England and Wales 2005) or section 51 of the Adults with Incapacity Act (Scotland 2000). The study was approved by the England and Wales Research Ethics Committee and the Scotland A Research Ethics Committee. A detailed description of the trial implementation has been published (citation removed for blind review).

Intervention

Nurses in both arms received training of three hours' duration focussed on the trial requirements and data collection. Nurses in sites assigned to the intervention arm received an additional three hours of training following guidelines laid out in the Learning Disability Epilepsy Specialist Nurse Competency Framework (Doherty et al. 2013). The framework outlines competencies considered by the developers of the framework to be central to effective performance in supporting participants with epilepsy and ID. These competencies range across the domains of clinical diagnosis and management of epilepsy, assessing and managing risk, impact of daily life on epilepsy, evidence-based practice, multidisciplinary team working, and professional development. In each domain a range of competencies are described such that the framework can be tailored to the experience level of each individual nurse using it,

according to their self-assessed competence level (‘novice’, ‘competent’ or ‘expert’). Nurses in both arms were free to practice as they judged clinically appropriate.

Assessments and data collection

Data from trial participants were collected by trial researchers at baseline and after six months, predominantly through telephone interview with the primary carer. Demographic data were collected at baseline, including postcode, which allowed assignment of the relevant Index of Multiple Deprivation (IMD) score. The IMD ranks localities according to measures of deprivation in multiple domains (Noble et al. 2006). Resource use was recorded at baseline and follow-up using a modified version of the Client Services Receipt Inventory which had been utilised in a previous study of the costs of supporting adults with epilepsy and intellectual disability (citation removed for blind review). The instrument reported the type of accommodation in which participants lived, their experience of respite care and holidays over the previous six months and contact with health and social care professionals related to their epilepsy over the previous month. Approximate contact times, the site of contact (home or clinic) and whether the family contributed to the cost (of social support) were recorded. Drug costs, investigations such as MRI scans and hospitalizations relevant to epilepsy were recorded. The instrument also reported the number of hours of informal care participants received. A copy of the questionnaire is included in the supplementary material.

Structured daily diaries were used by the nurses in the study to collect data describing the time spent by the nurses supporting participants. The diaries were completed on a daily basis and logged the duration of time spent in activities supporting people with epilepsy, the indication for the intervention and the type of activity.

Unit costs for each element of resource use were sought from appropriate national sources in 2014/2015 GBP (Curtis & Burns, 2015). Costs were inflated to 2014/2015 GBP where necessary using the Hospital and Community Health Services inflation index (Curtis & Burns, 2015). The following unit costs were obtained from Unit Costs of Health and Social Care 2015 (Curtis & Burns, 2015): outpatient appointment - £112; emergency ambulance - £231; passenger transport ambulance - £53; accommodation costs (classified as 4-bed house - £1,631 per week, 8-bed house - £1,284 per week; and supported living £906 per week); respite care - £280 per night (based on the mean cost for residential overnight respite care for disabled children), day care activities (such as adult education and social clubs) - £81 per session; hourly costs of primary health care providers, social workers, family support workers and home care support. Hourly costs of primary care providers are listed in table S1 in the supplementary material. A multiplier of 1.158 was applied to costs per contact hour for home visits; the multiplier is the ratio of NHS reference costs for home and clinic appointments with an occupational therapist (Department of Health & Social Care, 2015). Hourly costs were multiplied by 1.35 to estimate costs per patient contact hour based on costs per hour and patient contact hour for consultant surgeons reported in Unit costs of Health and Social Care 2010 (Curtis, 2010). Accident and Emergency admission costs (£103.67) and day hospital costs (£99.30) were derived from Unit Costs of Health and Social Care 2010 and inflated.

The cost of hospital inpatient stays was estimated at £400 per day based on data from the Department of Health (Department of Health, 2015). Drug costs were estimated on the basis of dose, frequency, formulation and brand, where stated, from data in the British National Formulary (British National Formulary, accessed November 2016). Costs of tests and investigations were taken from NHS Reference costs (Department of Health & Social Care, 2015). The cost of holidays was estimated according to duration

data matched to typical holiday packages by a specialist provider (JollyDays). A cost of £5.08 per 'meal on wheels' was derived from published sources (Banerjee et al. 2013). Costs for cleaning (£12 per hour) and laundry services (£14) were derived from online suppliers.

Informal care was valued at the gross market wage rate by category of employment (Office for National Statistics, 2016), or at the minimum wage for 2014/2015 of £6.50 per hour for unemployed or retired carers.

Costs for contacts with nurses were estimated according to duration costs per contact hour by Agenda for Change salary band (Curtis & Burns, 2015). Costs for home visits were multiplied by 1.158 as previously described. We did not include the costs of training nurses as it is unclear what proportion of such costs should be attributed to the support of participants over the first six months following training.

Analysis of costs

The cost analysis applied a health and social care perspective as recommended by NICE guidelines (NICE, 2013). A secondary analysis included costs falling on families, primarily travel and informal care but also paid social services to provide a societal perspective. The collection of resource use data over six months precluded the need for discounting. We undertook some ad hoc imputation of missing resource use data based on mean values for observed data where it was evident that contact with professionals had occurred but the duration was missing. Otherwise costs in the relevant category were recorded as missing when data were incomplete. Missing data were imputed using Multiple Imputation with Chained Equations in Stata (version 15) (Rubin, 2004). Predictive Mean Matching was used to replicate the distribution of observed data for missing data where the observed data was non-Normally distributed (Little, 1988). The imputation routine included generic (EQ-5D-5L, Devlin et al. 2018)

and epilepsy specific (ELDQOL) quality of life at baseline and follow-up, the level of ID (categorised as mild, moderate, severe or profound), sex, age, the number of Tonic-Clonic seizures (categorised), IMD quintile, and the site mean of self-assessed competence (novice, competent or expert) and workload of epilepsy nurse specialists in the trial (calculated as the site mean of the number of people with epilepsy per nurse).

The impact of the intervention on costs per participant recorded at six months was determined with adjustment for differences in baseline characteristics of participants and nurses. Each of the variables included in the imputation routine (listed above) were also included as covariates in the estimation of intervention costs. We did not envisage many participants with zero costs but we did expect the usual skewed data. The suitability of assuming Gaussian, Inverse Gaussian, Gamma and Poisson distributions for the dependent variable (cost) and an additive (linear) or multiplicative (log) link to the independent variables were explored through Generalised Linear Modelling (Blough & Ramsey, 2000). Uncertainty in the estimates of the impact of the intervention on costs were quantified through bootstrapping. One thousand bootstrap replicates were created prior to imputation of missing data using MI; treatment effects were estimated through regression modelling across imputed datasets for each bootstrap replicate with effects combined using Rubin's rules to generate a single value for each bootstrap replicate (Rubin, 2004). Non-parametric 95% confidence intervals were determined after ranking the values generated across the bootstrap replicates. All statistical analysis was undertaken in Stata version 15.

Subgroup analysis and sensitivity analysis

We pre-specified analysis according to the subgroups mild/moderate ID and severe/profound ID. Accommodation costs for participants in the formal care sector

were large and crudely estimated. Consequently, we undertook sensitivity analysis in which these costs were ignored. Our primary analysis allowed adjustment for differences in baseline characteristics but did not account for the clustering of data in the bootstrap routine. We undertook a sensitivity analysis which applied a two-stage bootstrap routine which explicitly accounted for clustering (Ng et al. 2013). Unfortunately, the routine did not support adjustment for differences at baseline. After undertaking MI, the two-stage bootstrap was applied to each of 20 imputed datasets. Mean cost differences were derived through combining the bootstrapped estimate of mean costs across the 20 imputed datasets using Rubin's rules (Rubin, 2004). Confidence intervals were estimated parametrically after deriving the overall standard deviation from bootstrapped standard error estimate for each imputed dataset using Rubin's rules.

Results

Raw data

Recruitment of sites began in September 2014 with the last site recruited a year later. Seventeen sites were recruited, of which eight were randomised to the competency framework intervention. Across the intervention sites 184 participants were recruited. Across the treatment as usual (TAU) sites 128 participants were recruited. A CONSORT diagram is available in the full report (citation removed for blind review). The primary reason for exclusion of participants was the lack of a seizure in the preceding six months (well controlled epilepsy). Table 1 compares the baseline characteristics of the participants and the specialist epilepsy nurses across treatment arms. There were notable differences in the experience level of nurses across the two arms; a higher proportion of nurses in the competency framework arm self-assessed their own competence level as 'expert', were nurse prescribers and were employed at

Agenda for Change band 8. A higher proportion of participants in the TAU arm had mild or moderate ID, were non-White and lived with family.

Table 2 presents the raw cost data at baseline and after six months and the proportion of missing data by treatment arm. Missing data at baseline amounted to 5% or less; missing data at follow-up was higher, but not more than 20% of the total. Formal accommodation costs amounted to around half the total costs falling on health and social care budgets, despite over a third of participants living with family. Drug costs, primary care costs, secondary care costs, respite care costs and costs of holidays were small. Social support, day care and informal care generated higher costs. Figure 1 contrasts costs at baseline and six months for all participants (data tabulated in table S2, supplementary material). The largest (absolute) change is seen in accommodation costs which increase at six months. These are accompanied by a reduction in social support costs. Health and social care and societal costs at baseline and six months were between 5% and 11% higher for participants with severe or profound ID.

Analysis of costs

Health and social care costs at six months were moderately skewed (Figure S1, supplementary material); examination of alternative assumptions regarding the distribution of the data and the dependence of the covariates using Generalised Linear Modelling did not identify a superior alternative to linear regression. Hence linear regression was used to adjust for baseline imbalances. Table 3 reports the difference in costs per participant at six months attributable to the competency framework and the non-parametric 95% confidence intervals. The competency framework is associated with a reduction in costs from both the health and social care and the societal perspective, albeit in each case the difference is not significant at the 5% level. 82% of

the bootstrap replicates (which capture sample uncertainty) indicate that the intervention is cost saving. The corresponding figure for societal costs is 93%.

Subgroup analysis suggested greater cost reductions in participants with severe or profound ID. When accommodation costs were excluded the reduction in health and social care costs associated with the intervention arm was slightly higher, but the likelihood that the intervention saved costs was unchanged. After applying MI for missing data prior to a two-stage bootstrap the estimated reduction in health and social care costs associated with the intervention is increased but confidence intervals still include zero.

Discussion

This is the first study internationally that explores the costs of nurses using a competency framework to support people with intellectual disabilities and epilepsy. Our findings indicate that the competency framework is unlikely to increase costs and may reduce the overall costs of caring for adults with epilepsy and ID. The competency framework may be streamlining decision making, enabling self-assessment of competence to practise, developing the nurses' scope of practice, and reducing duplication of care support (Halcomb et al. 2016). The trial was powered on the primary outcome and our data are insufficient to be certain that the competency framework lowered overall costs.

Our data illustrate the high cost of supporting adults with epilepsy and ID. Costs of accommodation and social support dwarf medical costs. These findings are consistent with previous studies that highlight the high social care costs of supporting people with epilepsy and ID (Morgan et al. 2003; Burke et al. 1999). Published evidence on the impact of specialist epilepsy nurses on costs is mainly focussed on secondary care.

Implementation of specialist epilepsy nurses have generated savings from reductions in emergency department admissions (Beasley, 2009; Warren et al. 1998), length of stay (Noble et al. 2014), or substitution of consultant led care (Johnson et al. 2010). Our data suggest the scope for cost savings in the provision of secondary care to this group is very limited. A quasi-experimental study across 14 GP practices in Bristol found no effect of specialist epilepsy nurses on resource use (Mills et al. 1999). None of the studies above addressed services specifically targeted at people with ID, and some excluded them (Noble et al. 2014). Our study is likely the largest of its kind with respect to people with epilepsy and ID, and our data suggest comorbid ID is associated with significant additional costs.

The sampling frame for the trial included seventeen sites across England, Scotland and Wales providing generalisability of the results across the UK and given the high prevalence of epilepsy in ID internationally, this study is of global relevance. We deliberately chose to recruit participants whose epilepsy was not sufficiently controlled to prevent seizures. Management of care is likely to be more challenging in this group providing greater scope for the competency framework to improve outcomes and possibly also to reduce costs. Whilst the scope for cost saving may be smaller in people with well managed epilepsy it seems unlikely that the trial findings would be reversed in this group. We took reasonable steps to reduce the impact of bias. Data were collected over the telephone by research assistants blinded to the allocation of the participant, and participants themselves and their carers were not explicitly informed about their allocation to treatment. We collected cost data using an instrument adapted from a previous study. We were aware of the limitations of the instrument with respect to accommodation costs; detailed appraisal of these costs would have required a level of study of accommodations that was impractical and difficult to justify. Whilst accommodation costs are high we did not expect the intervention to instigate a change

in these costs. In that respect the consistency of findings in the base case and sensitivity analysis is encouraging. The generalizability of our findings depends on the extent of similarities in health care between the UK and other settings. Further research in different health care settings might establish the reproducibility of our findings in different settings~~.~~, and explore the potential long-term benefits which may accrue from the continuing professional development element of the framework.

Our study has some limitations. We struggled to recruit the number of participants from each centre that we had originally envisaged. However, we were able to offset this by recruiting more sites. We did not find a significant reduction in costs associated with the competency framework; a larger trial may have been able to show a significant difference. We attempted to apply the most rigorous available statistical techniques to the analysis of our data, included a principled approach to missing data through the use of Multiple Imputation and a non-parametric approach to quantifying uncertainty which accommodated the skewed distribution of the cost data. However, we had to address the impact of clustering and the impact of baseline differences in separate analyses.

Conclusion

Current clinical guidelines recommend a role for epilepsy nurse specialists, particularly in the management of complex epilepsy (Kerr et al. 2014; NICE 2012). However, as noted in the Introduction, such nurses are a rare and relatively expensive resource. But nurses trained in intellectual disabilities are employed in most if not all community ID teams in the United Kingdom. The results of the (name removed for blind review) trial suggest that such nurses, working to the competency framework to help manage the epilepsy of adults with ID, are unlikely to increase the costs of supporting people with epilepsy and ID, and may reduce them. Thus, there may be a role for the framework in

enhancing the cost-effectiveness of support for adults with epilepsy and an ID, using the existing workforce. Our findings add to a modest but growing literature demonstrating the potential for nurse-led epilepsy care to reduce care costs.

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Tables

Variable	Category	Competency Framework		TAU		Overall	
		N	%	N	%	N	%
Nurses							
Compet. level[a]	Novice	0/18	0	8/50	16.0	8/68	11.8
	Expert	7/18	38.9	10/50	20.0	17/68	25.0
Prescriber[b]		5/17	29.4	1/45	2.2	6/62	9.7
Full time[c]		8/14	57.1	17/24	70.8	25/38	65.8
AfC Band[d]	5	2/15	13.3	11/46	23.9	13/61	21.3
	6	7/15	46.7	23/46	50.0	30/61	49.2
	7	3/15	20.0	11/46	23.9	14/61	23.0
Participants							
Male		99/184	53.8	61/128	47.7	160/312	51.3
Non-white		15/179	8.4	25/125	20.0	40/304	13.2
Level of ID[e]	Mild	19/173	11.0	21/107	19.6	40/280	14.3
	Moderate	31/173	17.9	24/107	22.4	55/280	19.6
	Severe	101/173	58.4	53/107	49.5	154/280	55.0
Accomm.[f]	Group	78/177	44.1	40/122	32.8	118/299	39.5
	Family	57/177	32.2	57/122	46.7	114/299	38.1
	Indep.	13/177	7.3	9/122	7.4	22/299	7.4
IMD	Most deprived	33/179	18.4	27/126	21.4	60/305	19.7

Accomm. – accommodation. AfC – Agenda for Change. Compet. - competence. IMD – Index of Multiple Deprivation. Indep. – independent. [a]Self assessed competency level according to the categories: novice, competent, expert. [b]Proportion of specialist epilepsy nurses who were nurse prescribers. [c]Specialist epilepsy nurses employed full time rather than part time. [d]Agenda for Change salary band of specialist epilepsy nurses (categories 5,6,7 and 8). [e]ID level categorised as mild, moderate, severe or profound. [f]Accommodation categorised as Group home, living with family, living independently, other.

Table 1. Characteristics of the study population

	Competency Framework (n = 184)				TAU (n = 128)			
	baseline		Follow-up		baseline		Follow-up	
	Mean (£)	Mis. (%)	Mean (£)	Mis. (%)	Mean (£)	Mis. (%)	Mean (£)	Mis. (%)
Drugs	160	0%	174	0%	200	0%	160	0%
Accommodation	3,853	4%	3,938	14%	2,949	5%	3,351	17%
Respite care	176	4%	153	14%	129	5%	165	19%
Holidays	71	4%	54	14%	70	5%	70	19%
Primary health	181	4%	162	14%	221	5%	244	19%
Social Care	795	4%	559	14%	859	5%	775	19%
Day care	996	4%	1,062	14%	980	5%	1,259	19%
Hospital visits	25	4%	54	14%	50	5%	93	19%
Participant/ family costs	208	0%	224	0%	156	0%	165	0%
Informal care	1,745	4%	1,783	14%	2,537	4%	2,652	19%
Epilepsy nurse	nc		57	0%	nc		57	0%
Total H&SS[a]	6,276	4%	6,247	14%	5,470	5%	6,288	19%
Total societal	8,238	4%	8,288	14%	8,191	5%	9,142	20%

[a]H&SS – Health and Social Services; nc – not collected

Table 2. Raw monthly cost data at baseline and follow-up by treatment arm.

	Mean	95% confidence interval[a]
Health & social care costs (£)	-357	-986 to 294
Societal costs (£)	-631	-1,473 to 181
Health & social care costs, mild/moderate ID (£)	-221	-1,209 to 732
Health & social care costs, severe/profound ID (£)	-457	-1,286 to 414
Societal care costs, mild/moderate ID (£)	-312	-1,682 to 1,002
Societal care costs, severe/profound ID (£)	-864	-1,953 to 160
Health & social care costs excluding accommodation (£)	-535	-1,057 to 238
Health & social care costs two stage bootstrap (£)	-588	-1879 to 703

[a]Non-parametric interval derived from ranked bootstrap replicates with the exception of the confidence interval for the two-stage bootstrap which was derived from overall standard deviation of the bootstrapped mean costs across 20 imputed datasets.

Table 3. Impact of the competency framework on monthly costs.

Figure legends

Figure 1. Costs by category at baseline and follow-up for all participants.